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Sensory Features and Repetitive Behaviors in Children with Autism and Developmental Delays

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Abstract

This study combined parent and observational measures to examine the association between aberrant sensory features and restricted, repetitive behaviors in children with autism ($N = 67$) and those with developmental delays ($N = 42$). Confirmatory factor analysis was used to empirically validate three sensory constructs of interest: hyperresponsiveness, hyporesponsiveness, and sensory seeking. Examining the association between the three derived sensory factor scores and scores on the Repetitive Behavior Scales—Revised revealed the co-occurrence of these behaviors in both clinical groups. Specifically, high levels of hyperresponsive behaviors predicted high levels of repetitive behaviors, and the relationship between these variables remained the same controlling for mental age. We primarily found non-significant associations between hyporesponsiveness or sensory seeking and repetitive behaviors, with the exception that sensory seeking was associated with ritualistic/sameness behaviors. These findings suggest that shared neurobiological mechanisms may underlie hyperresponsive sensory symptoms and repetitive behaviors and have implications for diagnostic classification as well as intervention.

Keywords

autism; repetitive behaviors; responses to sensory stimuli; sensory symptoms

Introduction

Autism is a neurodevelopmental disorder highly associated with genetic risk [Dawson, 2008]. The core symptoms of autism include impaired social reciprocity, nonverbal and verbal communication, and excessive displays of a restricted and repetitive range of behaviors and interests [ICD-10, World Health Organization, 1993; DSM-IV-TR, American Psychiatric Association, 1994]. Sensory features in autism often have co-occurred with the behavioral presentation of these established core symptoms. Sensory symptoms are considered secondary or associated features of the disorder because they have not been universally found in individuals with autism. Still there has been some discussion as to whether sensory symptoms in autism should be considered primary impairments that have resulted in problems relating to and interacting with others, particularly because for the developing child most of the world is explored and experienced via sensory input [Baranek, Parham, & Bodfish, 2005; Iarocci & McDonald, 2006; Rogers & Ozonoff, 2005].

Much of the confusion about the relationship between sensory symptoms and core features of autism has centered on the diagnostic category of restricted, repetitive behaviors and interests. Repetitive behaviors are defined by their repetition, inappropriateness, topographical similarity across contexts, and an overarching behavioral rigidity [Boyd, McBee, Holtzclaw, Baranek, & Bodfish, 2009]. The category of repetitive behaviors has included such behaviors as motor stereotypies (e.g., hand-flapping) and self-injury, which are presumed to provide intrinsic sensory stimulation to individuals with autism [Lovaas, Newsome, & Hickman, 1987]. Thus, part of the confusion may have simply stemmed from how difficult it has been to properly define and measure sensory symptoms in autism and parse those apart from repetitive behaviors. Yet, this is important because of the implications a relationship, or lack thereof, between sensory symptoms and repetitive behaviors potentially has for diagnostic criteria, pathogenesis, and treatment strategies in autism. Strong relationships (as indicated by a high degree of co-occurrence) may suggest that additional descriptions of sensory symptoms should be added to the diagnostic criteria for autism to better characterize the clinical phenotype. Further, robust linkages may indicate shared neurobiological mechanisms underlie these behavioral features, or that similar intervention approaches could be used to treat both classes of behaviors. In contrast, weak relationships may suggest these behaviors are more distinct than previously thought and that differential treatment approaches and taxonomies are needed, possibly because of dissimilar pathogenesis.

Three sensory patterns have emerged in the autism research literature: hyperresponsiveness (i.e., behavioral over-reactivity to sensory stimuli); hyporesponsiveness (i.e., behavioral under-reactivity to sensory stimuli); and sensory seeking (i.e., craving/fascination with certain stimuli). These categories are based on a combination of the proposed nosology for sensory processing disorders [Ben-Sasson et al., 2009; Miller, Anzalone, Lane, Cernak, & Osten 2007], and empirical findings in autism [Baranek, David, Poe, Stone, & Watson, 2006; Ben-Sasson et al., 2009; Liss, Saulnier, Fein, & Kinsbourne, 2006]. Little is known about the relationship of these patterns to each other as well as core symptoms of autism. For example, researchers have found that hyporesponsiveness and hyperresponsiveness co-occurred in autism [Baranek et al., 2006; Ben-Sasson et al., 2009; Greenspan & Weider, 1997; Hirstein, Iversen, & Ramachandran, 2001; Williams, 1994]. Using a parent report measure, however, Baranek et al. [2006] found the *hyper* responsive sensory pattern to be similar in autism and developmentally delayed (DD) samples, yet, this sensory type discriminated both of these groups from the typical comparisons. Follow-up studies that included observational measures have confirmed these findings and demonstrated that mental age (MA) was a strong predictor of hyperresponsiveness [Baranek, Boyd, Poe, David, & Watson, 2007]. In comparison, Baranek et al. [2006] found that sensory *hypo* responsiveness discriminated the autism group from both the DD and typical comparison groups, thus underscoring the importance of studying under-responsiveness as well as over-responsiveness to sensory stimuli. Children with autism also display higher frequencies of sensory seeking behaviors. In previous studies, it was reported that toddler- and preschool-aged children with autism differed from their typically developing peers in the excessive display of under-reactive behaviors, hypersensitivities, and sensory seeking behaviors [Ben-Sasson et al., 2008; Rogers, Hepburn, & Wehner, 2003; Watling, Dietz, & White, 2001]. A meta-analysis conducted by Ben-Sasson et al. [2009] found the behavioral presentation of this triad of sensory symptoms in autism to be moderated, in general, by (a) chronological age (CA) of individuals included in the study (larger effect sizes found for children up to 9 years of age vs. older children), (b) number of children with autism included in the study (larger effect sizes when >80% of sample had a diagnosis of autism), and (c) whether groups were matched on MA vs. CA (larger effect sizes for CA matched groups). At present the compilation of evidence underscores the importance of investigating the three patterns of sensory symptoms in autism to fully understand any relationship with core deficit areas as

well as the role developmental maturation plays in the expression of sensory and repetitive behavior symptoms.

In two prior studies, researchers reported associations between sensory features and repetitive behaviors in autism [Boyd et al., 2009; Gabriels et al., 2008]. Gabriels et al. used parent report measures to examine the association between these atypical behaviors in a sample of 70 children and adolescents with autism spectrum disorders. The researchers found consistently high levels of sensory features and repetitive behaviors in a subgroup of the sample. Similarly, Boyd et al. found the same relationship in a group of school-aged children (6–17 years of age) with high-functioning autism. These studies demonstrated that sensory symptoms and repetitive behaviors often co-occurred in autism. However, both studies only used parent report measures of sensory and repetitive behaviors. Observational methods are needed to validate these clinical phenomena and their interrelationships. Further, neither study investigated if there were differential relationships between sensory features and repetitive behaviors based on the sensory pattern(s) the child displayed. Examining any differential relationships between the expression of specific types of sensory symptoms and repetitive behaviors provides a more comprehensive understanding of a potential sensory phenotype in autism.

In summary, little is known about the association between patterns of sensory features and repetitive behaviors in autism. Conceptually and empirically validated classification systems of sensory patterns (i.e., hyporesponsiveness, hyperresponsiveness, sensory seeking) should be examined to better inform differential diagnosis, pathogenesis, and treatment in children with autism and related disorders. The purpose of this study was to examine the association between sensory patterns and repetitive behaviors in young children with autism and developmental delays. Our specific research questions were: (1) What is the association between three empirically derived sensory constructs (i.e., hyperresponsiveness, hyporesponsiveness, and sensory seeking) and repetitive behaviors (i.e., compulsions, restricted interests, rituals/sameness, self-injury, and stereotypy) in children with autism and DD? and (2) Does any relationship differ as a function of the child's MA or clinical group status?

Methods

Participants

Recruitment—Two groups of participants were included in this study: children with autistic disorder (AD) ($N = 67$) or developmental delay (DD) ($N = 42$). See Table 1 for a summary of participant and family descriptive and demographic information. The families received monetary incentives (\$25–\$75 dependent upon their child's age and diagnostic status) for their participation in the study which included standardized developmental assessments, and various sensory processing measures. The children also received a small toy or book for their contributions. We recruited participants through a variety of methods, including but not limited to, developmental evaluation clinics, parent support groups, and public schools in the state of North Carolina. In addition, the University of North Carolina (UNC) Neurodevelopmental Disorders Research Center Subject Registry was used to recruit families of children with autism. Once interested participants were identified, project staff contacted them via telephone, screened all participants for eligibility, and scheduled on-site assessments. Prior to administration of observational assessments, research staff screened children to confirm normal or corrected normal vision and hearing functions as atypical hearing or vision could impact sensory processing and subsequent behavioral reactions. UNC's Institutional Review Board approved the research.

Diagnostic confirmation—The AD group was comprised of children diagnosed with AD by an independent licensed psychologist or physician. We used the Autism Diagnostic Interview-Revised [Le Couteur, Lord, & Rutter, 2003] and Autism Diagnostic Observation Schedule (ADOS) [Lord, Rutter, DiLavore, & Risi, 1999] to confirm diagnosis. The DD group included three types of children: (a) those with known developmental disabilities and genetic syndromes (i.e., Down or Williams syndrome) associated with mental retardation ($N = 18$); (b) those with idiopathic developmental delay of a nonspecific nature based on either a global cognitive assessment (i.e., IQ >2 standard deviations below the mean), or significant delays (>1.5 standard deviations below the mean) in at least two developmental domains ($N = 20$) (i.e., Expressive Language, Receptive Language, Cognitive/Visual Reception, Fine or Gross Motor, and/or Adaptive Behavior); or (c) developmental delay that resulted from premature birth ($N = 4$). Children with idiopathic DD have been difficult to recruit; thus, children with known developmental disabilities or a delay that resulted from prematurity were included to augment sample size. We excluded children from the DD group if they had a clinical diagnosis of autism or a score above the autism cutoff on the Childhood Autism Rating Scale [CARS; Schopler, Reichler, & Renner, 1988] and/or ADOS. Exclusion criteria for both groups (AD and DD) were the presence of co-morbid conditions of autism, such as Fragile $\times$ syndrome and tuberous sclerosis; MA <6 months; cerebral palsy; uncorrected visual or hearing impairment; seizure disorder; or current receipt of psychopharmacological treatments as indicated by parents or from medical records.

Measures

Cognitive measures—The primary cognitive measure was the Visual Reception (VR) scale of the Mullen Scales of Early Learning [Mullen, 1995]. The Mullen is a standardized, child-administered measure of cognitive functioning for children from birth up to 68 months of age. The VR scale primarily tests visual discrimination and visual memory skills, and was selected as an efficient and valid measure of nonverbal cognitive performance for this study. Pearson correlations between the VR scale T score and the Early Learning Composite (overall standard score) on the Mullen were very high ($r = 0.96$). We utilized the age equivalent scores as a measure of MA. Some participants (13%) were above the age range recommended for the MSEL; thus, the Leiter International Performance Scale-Revised [Roid & Miller, 1997] was used in those cases (i.e., Leiter Brief IQ score converted to MA).

Repetitive behavior measure—The outcome measure was the Repetitive Behavior Scales—Revised [RBS-R; Bodfish, Symons, & Lewis, 1999], an informant-based rating scale of repetitive behavior (scores range from 0 = behavior not does not occur to 3 = behavior occurs and is severe). The RBS-R has six subscales that measure type/severity of a variety of repetitive behaviors in autism (i.e., compulsions, restricted interests, rituals, sameness, self-injurious behaviors, and stereotypy). Lam and Aman [2007] conducted a factor analysis of the RBS-R (based on $N = 320$ caregivers of individuals with autism) which generated a five factor solution with rituals and sameness loading on the same factor. Factor loadings for the RBS-R ranged from 0.51–0.66, accounting for 47.5% of the variance; internal consistency of the scales ranged from 0.78–0.91 and inter-rater agreement ranged from 0.57–0.73 [see Lam & Aman, 2007]. We used the five empirically derived factor scores in this study.

Sensory measures—A total of four sensory measures were used—two parent report and two observational—providing a multi-trait, multi-method approach to measuring the three sensory patterns of interest.

The Sensory Experiences Questionnaire [SEQ; Baranek, 1999a; Baranek et al., 2006] is a 43-item caregiver report questionnaire that measures frequencies of the child's unusual

sensory reactions across sensory modalities, contexts and response patterns (i.e., hypo/hyperresponsiveness, and sensory seeking). The Sensory Profile [SP; Dunn, 1999] is a 125-item questionnaire (Likert 5-point scale) that also measures sensory processing across sensory modalities (e.g., visual, tactile, auditory) in the context of daily activities. The Sensory Processing Assessment for Young Children [SPA; Baranek, 1999b] is a 20-minute semi-structured play-based, observational assessment that provides behavioral presses for these same sensory response patterns. The Tactile Defensiveness and Discrimination Test—Revised [TDDT-R; Baranek, 1998; Baranek & Berkson, 1994] is a 15-minute behavioral observation assessment of tactile processing (hyperresponsiveness and discrimination) for children with autism and other developmental disabilities.

Derivation of Sensory Construct Scores

Sensory construct scores—To measure the triad of sensory features in autism, items from each of the four sensory measures (i.e., SEQ, SP, SPA, and TDDT-R) were rigorously evaluated using a combined conceptual and empirical approach to validate the three sensory constructs of interest (i.e., hyper/hyporesponsiveness and sensory seeking). First, the study team established content validity to ensure that all items included in the final analyses to measure the sensory phenomena were in conceptual agreement [e.g., Beck & Gable, 2001; Grant & Davis, 1997], and then used a confirmatory factor analysis for empirical validation. Content validity was established by a team of three Ph.D. researchers and one doctoral student who were experts in the clinical manifestations of sensory processing features in autism, and they followed established guidelines to conduct an exhaustive review of all items [e.g., Blue, Marrero, & Black, 2008; Fitzner, 2007; Gould, Moore, McGuire, & Stebbins, 2008]. The process entailed two steps: (a) the researchers were divided into two pairs and each pair categorized items for two of the four assessments (to ensure that each assessment was reviewed by at least two evaluators) with each evaluator categorizing items independently; and (b) evaluators compared results and resolved any disagreements via a consensus across evaluators. Results of this process were consistent with the original conceptual models outlined by the primary authors of the instruments. We deleted sensory items that fell into three categories: (a) item was analogous to a repetitive behavior item on the RBS-R, which would have potentially conflated the measures (e.g., “rigid rituals in personal hygiene”); (b) item did not specifically address a sensory symptom (e.g., “weak grasp” indicated a motor problem that may or may not have a sensory basis); or (c) item could not be classified into a mutually exclusive category because it reflected more than one sensory construct (e.g., “prefers long sleeves” could indicate hyperresponsiveness or sensory seeking). Thus, the SEQ contributed a total of 30 items, the SP contributed 55 items, the TDDT contributed 25 items, and the SPA contributed 27 items.

Scores were transformed to ensure each item was scored on a 5-point scale with concordant valence across all four assessments (score of 1 being least severe sensory symptoms and a score of 5 being most severe). A mean score was derived for each sensory construct on each separate assessment. The items from each assessment were then entered into a confirmatory factor analysis to establish empirical validity of the three conceptually validated constructs of interest.

Confirmatory factor analysis—Confirmatory factor analytic procedures were used to generate scores for hyporesponsiveness, hyperresponsiveness, and sensory seeking for each of the measures. We created a measurement model in MPLUS (version 5) which had 5 latent constructs and 11 manifest variables (see Fig. 1). The 11 manifest variables came from the four different measures of sensory features—two parent (SEQ and SP) and two observed (SPA and TDDT-R). Three of the sensory instruments contributed hyporesponsiveness, hyper-responsiveness, and seeking items; the TDDT-R only contributed

hyperresponsiveness and seeking items. The inclusion of measures from two sources (parent and observer) introduces error into the model attributable to the format of the assessments. That is, we anticipated that there would be covariance among the parent measures and among the observational measures. To correct for potential rater effects, latent variables representing parent and observational measures also were included in the model. These method factors allowed us to control for their effects in estimating the three latent variables of interest. The measurement model had good fit, according to the fit indices, as evidenced by the non-significant chi-square ($\chi^2 = 32.4$; $P = 0.40$), the CFI near 1 (CFI = 0.997), and the RMSEA <0.08 (RMSEA = 0.021).

Statistical Analysis

Regression models—Once the factor analytic model was derived, each of the resulting sensory factor scores was regressed on the RBS-R scores to assess the associations between repetitive behaviors and sensory features. The RBS-R scores had a censored normal distribution because of an overabundance of scores of zero, and to account for this a series of repeated measures censored regression models were fit to the data using the xtobit command in STATA 10.0. The tobit model is appropriate for distributions where the dependent variable evidences a clustering at some lower limit [McDonald & Moffitt, 1980]. The percentage of children in the AD and DD groups that had mean scores of zero on a RBS-R subscale were 11% for stereotypies, 38% for self-injurious behaviors, 30% for compulsions, 17% for ritualistic/sameness behaviors, and 27% for restricted interests.

For the first set of analysis models, a separate model was fit for each of the three sensory constructs with hyperresponsiveness, hyporesponsiveness, and seeking factor scores entered into individual models as predictors. For this analysis each participant had five observations, one for each of the five RBS-R factor-derived subscales. Clinical group (AD or DD), gender, and MA were controlled for in these models. The models also included a categorical variable that designated the RBS-R subscale; interactions between this variable and the sensory factor score tested whether the association between the sensory construct and the RBS-R score varied between the subscales. Interactions also were evaluated to test whether this association varied as a function of MA or clinical group.

To test the independence of the effects of each sensory construct, a second analysis was performed to examine the association between the sensory factor scores and the RBS-R subscale scores. In this model, all three sensory factor scores were simultaneously entered into the model and regressed on the RBS-R scores, which allowed us to test the effects of one sensory construct while controlling for the effects of the other two constructs. This model also controlled for MA, clinical group, and gender.

Results

The means and standard deviations for the sensory constructs and RBS-R subscale scores are presented in Table 2. We found that the AD group had significantly higher scores than the DD group on the three sensory constructs and most subscales of the RBS-R except for self-injury. Pearson correlation coefficients were used to examine the intercorrelations among RBS-R subscales as well as the associations among the three sensory constructs. For the sensory constructs, hyporesponsiveness was moderately correlated with hyperresponsiveness ($r = 0.32$, $P = 0.0006$) and sensory seeking ($r = 0.34$, $P = 0.0003$); however, seeking and hyperresponsiveness were not significantly correlated ($r = 0.07$, $P = 0.484$). As indicated in Table 3, all RBS-R subscales were significantly correlated with each other.

Individual Models

A separate model was fit for each of the three sensory constructs with hyperresponsiveness, hyporesponsiveness, and seeking factor scores entered into individual models to examine their association with repetitive behaviors. In general, higher hyperresponsive behaviors were correlated with higher levels of repetitive behaviors in the AD and DD groups. Specifically, significant correlations were found between hyperresponsiveness and the presence of stereotypes, compulsions, and rituals/sameness behaviors. For sensory seeking, a significant correlation was only found for ritualistic/sameness behaviors, although correlations for stereotypes and self-injury approached significance. In addition, this association between the hyper and seeking constructs and repetitive behavior did not vary by clinical group (all $P > 0.15$). There also was no evidence that the association between repetitive and sensory behaviors significantly differed as a function of MA (all $P > 0.09$).

Simultaneous Models

A second model was fit to examine the association between each sensory construct and repetitive behaviors while controlling for the effect of the other two sensory constructs in the same model. For the simultaneous models, the associations between hyperresponsiveness or sensory seeking features and repetitive behavior remained the same as those found for the individual models. The relationship between repetitive behaviors and the two sensory features did not vary between the groups (all $P > 0.15$), and there was no evidence that the relationship differed by MA (all $P > 0.09$). The results for the individual and simultaneous models can be found in Table 4. Finally, the main effect for hyporesponsiveness and repetitive behaviors was not statistically significant for either the individual or simultaneous models (all $P > 0.10$).

Discussion

Using a combination of observational and parent report measures of sensory symptoms, significant associations were found between the hyperresponsive sensory construct and the presence of repetitive behaviors in children with autism and those with developmental delays. Thus, higher hyperresponsive scores were related to a variety of repetitive behaviors in both clinical groups; however, primarily non-significant associations were found between sensory seeking (craving/fascination with sensory stimuli) or hyporesponsiveness (behavioral under-reactivity to sensory stimuli) and repetitive behaviors. The one exception was the significant association found between sensory seeking and ritualistic/sameness behaviors. Preliminary findings suggest that sensory hyporesponsiveness and sensory seeking may be more associated with deficits in social-communication skills, at least in children with autism [Gay, Watson, Baranek, Poe, & Boyd, 2008]. In addition, the associations between sensory features and repetitive behaviors did not vary between the clinical groups. However, children with autism in our sample did have significantly higher levels of the three types of sensory patterns and most types of repetitive behavior in comparison to the group of children with DD.

Further, the association between sensory features and repetitive behaviors remained the same (i.e., higher hyper scores were associated with higher repetitive behavior scores) after controlling for participants' MAs. It is important to highlight that previous research has suggested that both sensory features and repetitive behaviors are independently related to the cognitive abilities or developmental maturation of individuals with autism. For example, Baranek et al. [2007] reported that MA was a predictor of hyperresponsiveness in children with autism and those with DD. With repetitive behaviors, it has been found that individuals with lower IQ levels, in particular nonverbal IQ, displayed more types of repetitive behaviors [Bishop, Reichler, & Lord, 2006; Bodfish, Symons, Parker, & Lewis, 2000].

Thus, developmental or cognitive maturation was predictive of the actual expression of these behaviors in children even though this variable may not have played a significant role in how these behaviors were related to one another. Other shared mechanisms may be responsible for their co-existence. A study by Gabriels et al. [2008] provided support for this finding. Those researchers also found that the actual relationship between abnormal sensory responses and repetitive behaviors was not affected by the IQ levels of the participants with autism, although their participants were older in age than our study sample (mean age = 10.8 years), and the researchers did not explore the different patterns of sensory features (i.e., hyper, hypo, and seeking). Understanding the relationship between these behaviors has both theoretical and clinical implications for the role of shared neurobiological mechanisms and treatment development.

While the present study did not directly test for physiological or biological correlates of these behaviors, previous research allows us to make theoretical postulations about the mechanisms underlying their relationship. First, it is important to highlight that research links both abnormal sensory features and repetitive behaviors to neuropathological mechanisms occurring early in development, which result in atypical sensory processing (e.g., long range underconnectivity in the cerebral cortex; and/or lack of “functional” connectivity) [Gomot, Belmonte, Bullmore, Bernard, & Baron-Cohen, 2008; Tommerdahl, Tannan, Holden, & Baranek, 2008] or aberrant restricted/repetitive behaviors (e.g., structural or functional differences in the caudate nucleus or frontal lobe) [Langen, Durston, Staal, Palmen, & van Engeland, 2007; Shafritz, Dichter, Baranek, & Belger, 2008; Thakkar et al., 2008]. We found a specific association between hyperresponsiveness and compulsive or ritualistic/sameness behaviors in both the individual and simultaneous data analysis models. Based on behavioral and physiological research on obsessive-compulsive disorder (OCD), a neurodevelopmental disorder that shares pathogenic and phenomenological similarities with autism [Rapoport & Inoff-Germain, 2000], researchers have found that anxiety plays a major role in the expression of compulsive/ritualistic behaviors [Abramowitz, Whiteside, & Deacon, 2005; Aouizerate et al., 2004; Piacentini & Langley, 2004]. Individuals with OCD often engage in these behaviors to alleviate anxiety that reoccurs as a result of obsessive thoughts. Hyperawareness of, or hypersensitivity to, stimuli in the environment (e.g., tags on the back of a child's shirt) may lead to similar feelings of anxiety, and subsequent engagement in compulsive or ritualistic behaviors to alleviate those feelings.

In typical development, researchers have speculated that ritualistic or OCD-like behaviors serve as compensatory mechanisms in fearful situations [Evans, Gray, & Leckman, 1999; Zohar & Felz, 2001], and allow individuals to establish predictability and control in a perceived chaotic environment [Fiske & Haslam, 1997; Zohar & Felz, 2001]. Compulsive and ritualistic types of repetitive behaviors in typically developing children begin to abate around six years of age [Evans et al., 1999]; yet, for children with developmental disorders these behaviors may continue to serve a compensatory strategy due to other cognitive capacities taking longer to come on-line, such as inhibitory control [Mosconi et al., 2009]. Further, at a biological level, some studies have linked the amygdala, which plays a role in regulating our reaction to environmental stimuli that are perceived as fearful or anxiety-provoking, to repetitive behavior in autism [Dziobek, Fleck, Rogers, Wolf, & Convit, 2006]. It is possible that the amygdala also plays a role in the expression of hyperresponsive sensory symptoms. We also found that hyperresponsiveness and stereotypy were correlated, and one of the long standing psychological theories has been that engagement in stereotypies served a homeostatic function and increased in situations associated with high levels of arousal [Turner, 1999]. In our study, sensory seeking was only significantly associated with ritualistic/sameness behaviors. This may indicate that there is a different pathogenesis associated with children who display hyperresponsiveness and ritualistic behaviors versus those who seek out certain stimuli in their environment but also display ritualistic/sameness

behaviors. Further research is needed to fully explicate the neurobiological underpinnings of our behavioral findings.

A multi-method, multi-trait approach to characterizing key sensory constructs, as used in this study, provides a more reliable and valid phenotypic characterization of sensory features that can be used as a basis for future experimental studies of pathogenesis in autism. We need additional research to address how co-presentation of aberrant sensory and repetitive behavior features relates to the child's adaptive functioning. It is plausible that a subgroup of children who present with high levels of both types of behaviors could have more trouble with everyday functioning in the home and community. Alternately, some theories [Baron-Cohen, Ashwin, Ashwin, Tavassoli, & Chakrabarti, 2009] postulate that hypersensitivities (i.e., enhanced sensory perception; lower sensory thresholds) may lead to greater attention to detail and thus facilitate hypersystemizing “talents” in high-functioning persons with autism. Thus, it is unclear to what extent sensory processing differences as found in this study may have adaptive vs. maladaptive effects on different individuals.

In conclusion, this study suggests that there is primarily a co-occurrence of hyperresponsive sensory features and repetitive behaviors in children with autism and DD, and these associations may be related to shared neurobiological mechanisms and have implications for diagnostic classification. Behavioral or educational treatments may need to address the co-presentation of these symptoms to maximize intervention effectiveness. Further research is needed to understand why the three sensory constructs are differentially related to core features of autism, and to determine neurobiological mechanisms that underlie these associations.

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References

- Abramowitz JS, Whiteside SP, Deacon BJ. The effectiveness of treatment for pediatric obsessive-compulsive disorder: A meta-analysis. *Behavior Therapy*. 2005; 36:55–63.
- American Psychiatric Association. *Diagnostic and statistical manual of mental disorders*. 4e.. American Psychiatric Publishing, Inc.; Washington, DC: 1994.
- Aouizerate B, Guehl D, Cuny E, Rougier A, Bioulac B, et al. Pathophysiology of obsessive-compulsive disorder: A necessary link between phenomenology, neuropsychology, imagery and physiology. *Progress in Neurobiology*. 2004; 72:195–221. [PubMed: 15130710]
- Baranek, GT. *Tactile Defensiveness and Discrimination Test-Revised (TDDT-R)*. University of North Carolina; Chapel Hill: 1998. Unpublished manuscript
- Baranek, GT. *Sensory Experiences Questionnaire (SEQ)*. University of North Carolina; Chapel Hill: 1999a. Unpublished manuscript
- Baranek, GT. *Sensory Processing Assessment for Young Children (SPA)*. University of North Carolina; Chapel Hill: 1999b. Unpublished manuscript
- Baranek GT, Berkson G. Tactile defensiveness in children with developmental disabilities: Responsiveness and habituation. *Journal of Autism and Developmental Disorders*. 1994; 24:457–471. [PubMed: 7961330]
- Baranek, GT.; Parham, DL.; Bodfish, JW. Sensory and motor features in autism: Assessment and intervention. In: Volkmar, F.; Paul, R.; Klin, A.; Cohen, editors. *Handbook of autism and pervasive developmental disorders*: Vol. 2. Assessment, interventions, and policy. 3e.. Wiley; Hoboken, NJ: 2005. p. 831-857.

- Baranek GT, David FJ, Poe MD, Stone WL, Watson LR. The Sensory Experiences Questionnaire: Discriminating response patterns in young children with autism, developmental delays, and typical development. *Journal of Child Psychology and Psychiatry*. 2006; 47:591–601. [PubMed: 16712636]
- Baranek GT, Boyd BA, Poe M, David F, Watson LR. Hyperresponsive sensory patterns in young children with autism, developmental delay, and typical development. *American Journal of Mental Retardation*. 2007; 112:233–245. [PubMed: 17559291]
- Baron-Cohen S, Ashwin E, Ashwin C, Tavassoli T, Chakrabarti B. Talent in autism: Hyper-systemizing, hyper-attention to detail and sensory hypersensitivity. *Philosophical Transactions of the Royal Society of Biological Sciences*. 2009; 364:1377–1383.
- Beck CT, Gable RK. Ensuring content validity: An illustration of the process. *Journal of Nursing Measurement*. 2001; 9:201–215. [PubMed: 11696942]
- Ben-Sasson A, Hen L, Fluss R, Cermak S, Engel-Yeger B, Gal E. A meta-analysis of sensory modulation symptoms in individuals with autism spectrum disorders. *Journal of Autism and Developmental Disorders*. 2009; 39:1–11. [PubMed: 18512135]
- Ben-Sasson A, Cermak S, Orsmond G, Tager-Flusberg H, Kadlec M, Carter A. Sensory clusters in toddlers with autism spectrum disorders: Differences in affective symptoms. *Journal of Child Psychology and Psychiatry*. 2008; 49:817–825. [PubMed: 18498344]
- Bishop S, Reichler J, Lord C. Association between restricted and repetitive behaviors and nonverbal IQ in children with autism spectrum disorders. *Child Neuropsychology*. 2006; 12:247–267. [PubMed: 16911971]
- Blue CL, Marrero DG, Black DR. Physical activity belief scales for diabetes risk: Development and psychometric testing. *Health Education & Behavior*. 2008; 35:316–331. [PubMed: 17620669]
- Bodfish JW, Symons FJ, Lewis MH. The Repetitive Behavior Scales (RBS). Western Carolina Center Research Reports. 1999
- Bodfish JW, Symons FJ, Parker DE, Lewis MH. Varieties of repetitive behavior in autism: Comparison to mental retardation. *Journal of Autism and Developmental Disabilities*. 2000; 30:237–243.
- Boyd BA, McBee M, Holtzclaw T, Baranek GT, Bodfish JW. Relationships among repetitive behaviors, sensory features, and executive functions in high functioning autism. *Research in Autism Spectrum Disorders*. 2009; 3:959–966.
- Dawson G. Early behavioral intervention, brain plasticity, and the prevention of autism spectrum disorder. *Development and Psychopathology*. 2008; 20:775–803. [PubMed: 18606031]
- Dunn, W. The sensory profile. Psychological Corporation; San Antonio, TX: 1999.
- Dziobek I, Fleck S, Rogers K, Wolf OT, Convit A. The ‘amygdala theory of autism’ revisited: Linking structure to behavior. *Neuropsychologia*. 2006; 44:1891–1899. [PubMed: 16566949]
- Evans DW, Gray FL, Leckman JF. The rituals, fears, and phobias of young children: Insights from development, psychopathology, and neurobiology. *Child Psychiatry and Human Development*. 1999; 29:261–276. [PubMed: 10422351]
- Fiske AP, Haslam N. Is obsessive-compulsive disorder a pathology of the human disposition to perform socially meaningful rituals? Evidence of similar content. *The Journal of Nervous and Mental Disease*. 1997; 185:211–222. [PubMed: 9114806]
- Fitzner K. Reliability and validity: A quick review. *Diabetes Educator*. 2007; 33:775. [PubMed: 17925583]
- Gabriels R, Agnew J, Miller J, Gralla J, Pan, et al. Is there a relationship between restricted, repetitive, stereotyped behaviors and interests and abnormal sensory response in children with autism spectrum disorders? *Research in Autism Spectrum Disorders*. 2008; 2:660–670.
- Gay, E.; Watson, LR.; Baranek, GT.; Poe, M.; Boyd, B. Sensory processing patterns in relation to social communicative skills in young children with autism; Gatlinburg Conference on Research and Theory in Intellectual and Developmental Disabilities; San Diego, CA. 2008.
- Gould J, Moore D, McGuire F, Stebbins R. Development of the serious leisure inventory and measure. *Journal of Leisure Research*. 2008; 40:47–68.
- Gomot M, Belmonte MK, Bullmore ET, Bernard FA, Baron-Cohen S. Brain hyper-reactivity to auditory novel targets in children with high functioning autism. *Brain*. 2008; 131:2479–2488. [PubMed: 18669482]

- Grant JS, Davis LL. Focus on quantitative methods. Selection and use of content experts for instrument development. *Research in Nursing and Health*. 1997; 20:269–274. [PubMed: 9179180]
- Greenspan S, Weider S. Developmental patterns and outcomes in infants and children with disorders in relating and communicating: A chart review of 200 cases of children with autistic spectrum diagnoses. *Journal of Developmental and Learning Disorders*. 1997; 1:87–141.
- Hirstein W, Iversen P, Ramachandran VS. Autonomic responses of autistic children to people and objects. *Proceedings of the Royal Society of London B, Biological Sciences*. 2001; 268:1883–1888.
- Iarocci G, McDonald J. Sensory integration and the perceptual experience of persons with autism. *Journal of Autism and Developmental Disorders*. 2006; 36:77–90. [PubMed: 16395537]
- Lam KSL, Aman MG. The Repetitive Behavior Scale—Revised: Independent validation in individuals with autism spectrum disorders. *Journal of Autism and Developmental Disorders*. 2007; 37:855–866. [PubMed: 17048092]
- Langen M, Durston S, Staal WG, Palmen S, van Engeland H. Caudate nucleus is enlarged in high-functioning medication-naïve subjects with autism. *Biological Psychiatry*. 2007; 62:262–266. [PubMed: 17224135]
- Le Couteur, A.; Lord, C.; Rutter, M. *Autism Diagnostic Interview-Revised (ADI-R)*. Western Psychological Corporation; Los Angeles: 2003.
- Liss M, Saulnier C, Fein D, Kinsbourne M. Sensory and attention abnormalities in autism spectrum disorders. *Autism*. 2006; 10:155–172. [PubMed: 16613865]
- Lord, C.; Rutter, M.; Dilavore, P.; Risi, S. *The Autism Diagnostic Observation Schedule (ADOS)*. Western Psychological Corporation; Los Angeles: 1999.
- Lovaas OI, Newsome C, Hickman C. Self-stimulatory behavior and perceptual reinforcement. *Journal of Applied Behavior Analysis*. 1987; 21:281–289.
- McDonald JF, Moffitt RA. The uses of tobit analysis. *Review of Economics and Statistics*. 1980; 62:318–321.
- Miller LJ, Anzalone ME, Lane S, Cemek S, Osten E. Concept evolution in sensory integration: A proposed nosology for diagnosis. *American Journal of Occupational Therapy*. 2007; 61:135–140. [PubMed: 17436834]
- Mosconi MW, Kay M, D'Cruz A, Seidenfeld A, Guter S, et al. Impaired inhibitory control is associated with higher order repetitive behaviors in autism spectrum disorders. *Psychological Medicine*. 2009; 39:1559–1566. [PubMed: 19154646]
- Mullen, EM. *Mullen Scales of Early Learning (AGS Edition)*. Western Psychological; Los Angeles: 1995.
- Piacentini J, Langley AK. Cognitive-behavioral therapy for children who have obsessive-compulsive disorder. *Journal of Clinical Psychology*. 2004; 60:1181–1194. [PubMed: 15389618]
- Rapoport JL, Inoff-Germain G. Practitioner review: Treatment of obsessive-compulsive disorder in children and adolescents. *Journal of Child Psychology and Psychiatry*. 2000; 41:419–431. [PubMed: 10836672]
- Roid, GH.; Miller, LJ. *Leiter international performance scale revised*. Stoelting Co.; Wood Dale, IL: 1997.
- Rogers SJ, Ozonoff S. Annotation: What do we know about sensory dysfunction in autism? A critical review of the empirical evidence. *Journal of Child Psychology and Psychiatry*. 2005; 46:1255–1268. [PubMed: 16313426]
- Rogers SJ, Hepburn S, Wehner E. Parent reports of sensory symptoms in toddlers with autism and those with other developmental disorders. *Journal of Autism and Developmental Disorders*. 2003; 33:631–642. [PubMed: 14714932]
- Schopler, E.; Reichler, RJ.; Renner, BR. *The childhood autism rating scale*. Western Psychological Services; Los Angeles, CA: 1988.
- Shafritz KM, Dichter GS, Baranek GT, Belger A. The neural circuitry mediating shifts in behavioral response and cognitive set in autism. *Biological Psychiatry*. 2008; 63:974–980. [PubMed: 17916328]

- Thakkar KN, Polli FE, Joseph RM, Tuch DS, Hadjikhani N, et al. Response monitoring, repetitive behaviour and anterior cingulate abnormalities in autism spectrum disorders. *Brain*. 2008; 131:2464–2478. [PubMed: 18550622]
- Tommerdahl M, Tannan V, Holden J, Baranek GT. Absence of stimulus-driven synchronization effects on sensory perception in autism: Evidence for local underconnectivity? *Behavioral and Brain Functions*. 2008; 4:1–9. [PubMed: 18173840]
- Turner M. Annotation: Repetitive behaviour [sic] in autism: A review of psychological research. *Journal of Child Psychology and Psychiatry*. 1999; 40:839–849. [PubMed: 10509879]
- Watling RL, Dietz J, White O. Comparison of Sensory Profile scores of young children with and without autism spectrum disorders. *The American Journal of Occupational Therapy*. 2001; 55:416–423. [PubMed: 11723986]
- Williams, D. *Nobody nowhere*. Random House Publishing Group; New York: 1994.
- World Health Organization. *The ICD-10 classification of mental and behavioral disorders: Diagnostic criteria for research*. WHO; Geneva: 1993.
- Zohar AH, Felz L. Ritualistic behavior in young children. *Journal of Abnormal Child Psychology*. 2001; 29:121–128. [PubMed: 11321627]

Figure 1. Sensory Constructs Measurement Model. SPA = Sensory Processing Assessment; TDDT = Tactile Defensiveness and Discrimination Test—Revised; SEQ = Sensory Experiences Questionnaire; SP = Sensory Profile.

Table 1

Participant and Family Demographic Information

	AD	DD
CA months—Mean (SD)	51.69 (17.07)	49.45 (24.19)
MA months—Mean (SD)	31.97 (20.84)	32.4 (17.39)
<i>Gender</i>		
Female	11	19
Male	56	23
<i>Mother's education</i>		
10–11 years	1	1
HS graduate	12	3
Partial College	13	7
College degree	23	16
Graduate degree	12	14
Missing	6	1
<i>Race</i>		
Asian	0	1
Black	8	3
White	48	33
More than One Race	5	4
Missing	6	1

Table 2

Group Differences in Sensory Constructs and RBS-R Subscales

	AD		DD		<i>P</i>
	Mean	SD	Mean	SD	
<i>Sensory Factor Scores</i>					
Hyporesponsive	0.12	0.47	−0.19	0.35	0.005
Hyperresponsive	0.07	0.32	−0.11	0.35	0.044
Seeking	0.04	0.17	−0.07	0.14	<0.001
<i>RBS-R subscales</i>					
Stereotypy	8.64	5.06	3.14	3.41	<0.0001
Self-injury	2.24	2.67	1.62	3.91	0.3945
Compulsions	3.12	3.14	1.31	2.14	0.0022
Rituals	6.52	5.86	3.07	4.22	0.001
Restricted Interests	3.73	2.48	1.48	2.1	<0.0001

Note. Group differences controlling for MA and gender.

Table 3

Correlation Matrix for RBS-R Subscale Scores and Sensory Constructs

	RBS-R Compulsions	RBS-R Restricted Interests	RBS-R Rituals	RBS-R Self-injury	RBS-R Stereotypy
RBS-R	1	0.45	0.75	0.51	0.62
Compulsions		<0.0001	<0.0001	<0.0001	<0.0001
RBS-R	0.45	1	0.67	0.22	0.67
Restricted Interests	<0.0001		<0.0001	0.023	<0.0001
RBS-R	0.75	0.67	1	0.46	0.66
Rituals	<0.0001	<0.0001		<0.0001	<0.0001
RBS-R	0.51	0.22	0.46	1	0.40
Self-injury	<0.0001	0.023	<0.0001		<0.0001
RBS-R	0.62	0.67	0.66	0.40	1
Stereotypy	<0.0001	<0.0001	<0.0001	<0.0001	
	Hyporesponsive	Hyperresponsive	Seeking		
Hyporesponsive	1	0.32 0.0006	0.34 0.0003		
Hyperresponsive	0.32 0.0006	1	0.07 0.4841		
Seeking	0.34 0.0003	0.07 0.4841	1		

Note. Correlations are for combined AD and DD groups (N=109).

Table 4
Relationship between Sensory Construct Scores and RBS-R Scores Controlling for Gender, MA, and Group

	Simultaneous Models				Individual Models					
	B	SE	P	95% CI	B	SE	P	95% CI		
Hyporesponsive										
Main Effect	0.41	0.80	0.606	-1.16	1.98	1.33	0.80	0.097	-0.24	2.90
Stereotypy	1.14	1.05	0.279	-0.93	3.21	2.17	1.03	0.036	0.15	4.19
Self-injury	0.95	1.10	0.387	-1.20	3.09	1.66	1.07	0.121	-0.44	3.76
Compulsions	-0.15	1.09	0.893	-2.28	1.99	0.70	1.06	0.509	-1.38	2.78
Rituals	0.21	1.07	0.847	-1.89	2.30	1.60	1.04	0.124	-0.44	3.64
Res. Interests	-0.08	1.07	0.937	-2.18	2.02	0.54	1.05	0.609	-1.51	2.58
Hyperresponsive										
Main Effect	3.05	1.02	0.003	1.05	5.05	3.31	1.00	0.001	1.35	5.27
Stereotypy	3.40	1.35	0.012	0.76	6.04	4.00	1.30	0.002	1.44	6.55
Self-injury	2.02	1.40	0.150	-0.73	4.77	2.52	1.36	0.063	-0.14	5.19
Compulsions	3.50	1.41	0.013	0.73	6.27	3.43	1.36	0.012	0.76	6.10
Rituals	4.47	1.35	0.001	1.83	7.11	4.68	1.30	<0.001	2.14	7.23
Res. Interests	1.87	1.37	0.175	-0.83	4.56	1.93	1.33	0.147	-0.67	4.53
Sensory Seeking										
Main Effect	4.43	2.22	0.046	0.07	8.79	4.93	2.31	0.033	0.40	9.46
Stereotypy	4.92	2.94	0.094	-0.84	10.67	5.78	3.00	0.054	-0.10	11.66
Self-injury	5.40	3.01	0.073	-0.51	11.30	6.03	3.07	0.050	0.00	12.06
Compulsions	1.26	3.02	0.677	-4.65	7.17	1.58	3.07	0.608	-4.44	7.59
Rituals	5.92	2.97	0.046	0.10	11.73	6.49	3.02	0.032	0.57	12.41
Res. Interests	4.66	3.02	0.122	-1.25	10.57	4.78	3.08	0.120	-1.25	10.81

Note. The values in bold indicate statistical significance.